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Case Report

An odontogenic gingival epithelial hamartoma (OGEH) possibly derived from remnants of the dental lamina ("dental laminoma")

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Summary A case of the rare so-called odontogenic gingival epithelial hamartoma (OGEH) located to the anterior maxilla in a 35-year-old woman is presented. Histology showed a small, well-circumscribed epithelial, hamartomatous lesion located to the gingival lamina propria, and totally separated from the covering oral epithelium. The lesion was composed of multiple clusters of well-delineated, uniform-sized, polyhedral odontogenic epithelial cells several of which showed clear cell differentiation. Only two of the previously published nine cases of OGEH showed histological features comparable to those reported here.

Introduction

Recent studies have established evidence that certain odontogenic cysts, tumours and hamartomatous lesion are derived from remnants of odontogenic epithelium persisting in the jaws after the completion of human odontogenesis.[1] Available evidence points to two main sources of the epithelium from which the lesions may be derived: the *dental lamina* (rests of Serres) and the *epithelial root sheath* (rests of Malassez). It is widely held that upon disintegration of these structures, the majority of the resulting epithelial islands, strands and cell clusters persist within the jaw bones and gingival tissues throughout life as "resting" cellular elements. Stimuli, the nature of which is poorly understood, may at any time trigger the vital but seemingly inactive or "resting" cell clusters to proliferation, leading to lesions as mentioned above.

The aim of this report is to present a case believed to be an odontogenic gingival epithelial hamartoma (OGEH) of the anterior maxillary gingiva possibly derived from remnants of the dental lamina.

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Case report

A 35-year-old woman complained of a slowly enlarging lesion of the maxillary labial gingiva of the left central incisor region. Its presence was first noticed by the patient some 5 years prior to her visit. Initially, the lesion was asymptomatic, but during the recent six months the patient had experienced occasional slight pain or "irritation" in the area. The patient thought that the appearance of the lesion might have been provoked by "energetic toothbrushing". Nothing in her medical history was significant, and the patient was in excellent health.

Clinical examination disclosed a small "sausage-shaped", non-movable tissue mass of the maxillary labial, marginal gingiva above the left central incisor, the longest diameter measuring approximately 8 mm. The covering mucosa was focally erythematous with small superficial ulcerations, in other areas the mucosal surface was whitish (frictional keratosis-like). The lesion was fibrous on palpation and gave the impression of a "fibrous epulis". Intraoral radiographs of the incisor area demonstrated a slight horizontal loss of the facial alveolar bone margin but no intraosseous lesion was seen. With the patient under local anaesthesia, the gingival mass was excised. The operation specimen was fixed in 10% buffered formalin and send for histology. Sedative periodontal dressings were placed post-operatively, with a subsequent uneventful post-operative course and excellent healing.

Histopathology

The operation specimen was subjected to serial sectioning. When viewed at a low-power scanning level (Fig. 1), a well-circumscribed nodule could be seen in the lamina propria. All sections examined clearly demonstrated that the lesional tissue was totally separated from the covering parakeratinized, stratified squamous surface epithelium by a band of fibrosed connective tissue containing a few scattered lymphocytes. The surface epithelium showed moderate, simple hyperplasia. The entire nodule was surrounded by layers of compressed collagen fibres forming a pseudocapsule. The nodule was composed of numerous rounded nests containing clusters of epithelial cells of putative odontogenic origin.

At higher magnification each cluster of epithelial cells was surrounded by collagen fibres of varying caliber, clearly demonstrated using polarized light

Figure 1 Low-power view of OGEH showing lesional tissue separated from oral epithelium by fibrous pseudocapsule. H&E, ×50.

(Fig. 2). The stroma between the individual clusters was occupied by loose connective tissue containing thin-walled dilated vessels and fibroblasts, in other areas by fibrous tissue (Fig. 3). The polyhedral epithelial cells within the nests were of uniform-size with a basophilic rounded nucleus (Fig. 4). Several epithelial cells showed clear cell differentiation with vacuolated or clear cytoplasm that did not stain with mucicarmine or Alcian blue, whereas some cells were positive in periodic acid-Schiff stain (PAS, with or without diastase). Immunohistochemically, most epithelial cells in the nodules expressed slightly to moderate positive keratin antigens, but were negative for anti-S-100

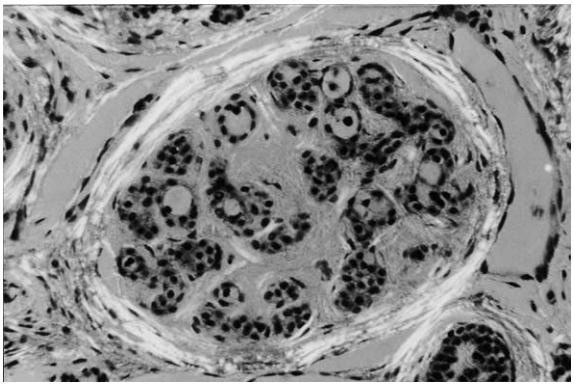

Figure 2 Cluster of epithelial cells surrounded by collagenous pseudocapsule. H&E/polarized light, ×150.

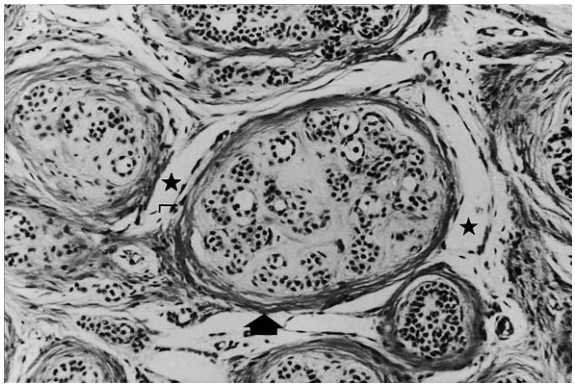

Figure 3 Group of epithelial cell clusters each bordered by fibrous pseudocapsule (arrow-head). In this area the connective tissue stroma is loose containing thin-walled dilated vessels (asterixs). H&E, ×120.

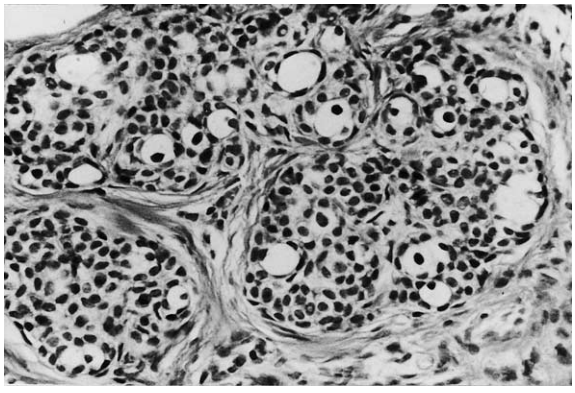

Figure 4 Epithelial cell clusters showing clear cell differentiation of several cells. H&E, ×150.

antibodies. The cell nests did not display a peripheral layer with a cuboidal or columnar shape and the nuclei were not polarized nor were cells appearing squamous with or without sign of keratinization encountered. No evidence of inductive reactions or hyalinisation was seen at the epithelial/mesenchymal interface. Mitotic figures and signs of cellular atypia were not present.

Diagnosis: Odontogenic gingival epithelial hamartoma (OGEH) of possible dental lamina origin ("dental laminoma").

Discussion

The above innocuous lesion proved on histology not to be that of an inflammatory fibrous hyperplasia ("epulis"), a lesion which on the initial oral examination seemed a likely clinical differential diagnosis. The presence of aggregations of clusters of epithelial odontogenic cells many of which showed clear cell differentiation, favoured a developmental aetiology reminiscent of a dental lamina origin. In the case presented here an inflammatory stimulus did not seem to have acted as a trigger mechanism.

Epithelial odontogenic hamartomas are considered to represent a transitional stage between a true epithelial odontogenic neoplasm and a tumour-like (in the sense of a new growth of tissue cells), but essentially non-neoplastic lesion. It is characterized by an abnormal mixture of cells or tissues native to the part, but in excess and unanticipated.[2] Hamartomas lack the diagnostic features of persistent and uncoordinated growth that characterize tumours.

The odontogenic gingival (extraosseous or soft tissue) epithelial hamartomas (OGEH) believed to originate from remnants of the dental lamina are very rare lesions with only 10 cases,[3–8] including the present one having been published to date (see Table 1, where cases nos. 8 and 10 are excluded due to the intraosseous involvement of the reported lesions). The eight cases where the age of the patients has been revealed, have all occurred in adults (mean age 53.4 years) and the majority of these were present in females (F:M = 7:1). The terminology—OGEH—was initially suggested by Baden and co-workers,[3,4] and has been applied by subsequent authors using various terms (see Table 1). Most cases summarized in Table 1, document lesions composed of more or less circumscribed islands and strands of odontogenic epithelium, some of which show a peripheral layer of cuboidal to columnar cells. In some cases, lesional epithelial cells appeared to be in direct continuity with the basal cell layer of the oral stratified squamous epithelium, features not encountered in the present case.

Among the previously reported nine cases, only two (Table 1, cases nos. 6 and 7) have—in some areas—demonstrated histological features that can be compared to those described in the present report.

Thus, the case presented here seems quite unique histologically. The most outstanding feature is the occurrence of multiple, well-delineated clusters of uniform odontogenic epithelial cells (several of which show clear cell differentiation), each cluster being surrounded by a distinct pseudocapsule. The accumulation of cell clusters are wrapped in a slightly thicker connective tissue pseudocapsule, totally separating the entire lesion from the basal cell layer of the oral epithelium.

From a differential diagnostic point of view, some recognized odontogenic lesions have to be

Table 1 Data from published cases of odontogenic gingival epithelial hamartoma (OGEH)

Case no.	Author(s)	Age (years)	Gender	Location[a]	Radiology	Size (mm)	Microscopical diagnosis
1	Baden et al.[3]	59	F	33 or 43	Neg.[b]	n.i.[c]	Hamartoma of dental lamina rests (odontogenic epithelial hamartoma)
2		55	M	13, 14	Neg.[b]	2.5 across	—
3		65	F	34, 35	Neg.[b]	n.i.	—
4	Baden and Splaver[4]	60	F	41, 42	Neg.[b]	3 across	OGEH
5	Gardner[5]	50	F	36—38	Neg.[b]	15×10 ×8	Hamartoma of dental lamina (peripheral odontogenic fibroma)
6	Sciubba and Zola[6]	40	F	31, 41	Neg.[b]	2×3	Odontogenic epithelial hamartoma
7	Moskow and Baden[7]	n.i.	n.i.	38 or 48	n.i.	n.i.	Gingival odontogenic hamartoma
8		n.i.	n.i.	16, 15 or 25, 26[d]	n.i.	n.i.	Epithelial odontogenic hamartoma (central type) with ameloblastoid features
9		n.i.	n.i.	Maxill. molar	n.i.	n.i.	Gingival odontogenic epithelial hamartoma with features of CEOT[e]
10		n.i.	n.i.	Maxill. gingiva and bone[d]			Odontogenic epithelial hamartoma (gingival and central type) with features of CEOT
11	Kitano et al.[8]	63	F	35	n.i.	n.i.	Odontogenic epithelial hamartoma of gingiva
12	Present case	35	F	21	Neg.[b]	10 across	Gingival epithelial hamartoma of dental lamina origin

n.i.: no information available.
[a] According to the FDI nomenclature.
[b] No bone involved.
[c] Described as "pea-size".
[d] Bone involved.
[e] Calcifying epithelial odontogenic tumour.

considered. Philipsen et al.[9] in their review article on the peripheral ameloblastoma (PA) argued that data on age, clinical findings including location, radiological features, behavioural pattern and histology may indicate that OGEH and at least some PAs could be considered one and the same lesion. Sciubba and Zola[6] suggested that the diagnosis *peripheral ameloblastoma* should be confined to those OGEH-lesions that originate from overlying mucosal epithelium and demonstrate infiltrative characteristics. A second lesion to consider is the peripheral odontogenic fibroma (POF, WHO-type or in accordance with the new WHO Classification of Tumours of the Head and Neck,[10] the cell-rich type). The proliferation of strands and islands of odontogenic epithelium in this tumour type may be so extensive as to make the distinction from both PA and OGEH very difficult if at all possible.

At this moment in time it seems likely that at least some reported cases of PAs, POFs (cell-rich type) and OGEHs may represent a histomorphological spectrum of a hamartomatous lesion, the true nature of which can not be determined until several more cases are available for evaluation.

References

1. Reichart PA, Philipsen HP. Odontogenic tumors and allied lesions. Berlin, London: Quintessence Publishing Co. Ltd.; 2004.
2. Gardner DG. The concept of hamartomas: its relevance to the pathogenesis of odontogenic lesions. *Oral Surg Oral Med Oral Pathol* 1978;**45**:884—6.

3. Baden E, Moskow BS, Moskow R. Odontogenic gingival epithelial hamartoma. *J Oral Surg* 1968;**26**:702—14.
4. Baden E, Splayer T. Odontogenic gingival epithelial hamartoma: report of a case. *J Oral Surg* 1973;**31**:932—5.
5. Gardner DG. An ameloblastoma and a hamartoma of the dental lamina in two siblings. *J Oral Surg* 1973;**31**:697—700.
6. Sciubba JJ, Zola MB. Odontogenic epithelial hamartoma. *Oral Surg Oral Med Oral Pathol* 1978;**45**:261—5.
7. Moskow BS, Baden E. Odontogenic epithelial hamartomas in periodontal structures. *J Clin Periodontol* 1989;**16**:92—7.
8. Kitano M, Landini G, Urago A, Okubo A, Mukai H, Yamashita S. Odontogenic epithelial hamartoma of the gingiva: a case report. *J Periodontol* 1991;**62**:452—7.
9. Philipsen HP, Reichart PA, Nikai H, Takata T, Kudo Y. Peripheral ameloblastoma: biological profile based on 160 cases from the literature. *Oral Oncol* 2001;**37**:17—27.
10. World Health Organization Classification of Tumours: Pathology and Genetics of Tumours of the Head and Neck. Odontogenic Tumours. Lyon: IARC Press; 2004, in press.